Gastric outlet obstruction caused by an ectopic pancreas in a neonate: A case report

著者	Fujii Shunsuke, Tanaka Hideaki, Ono Kentaro, Onizawa Yutaro, Miyazono Yayoi, Sakamoto Naoya, Gotoh Chikashi, Urita Yasuhisa, Shinkai Toko, Takayasu Hajime, Sumazaki Ryo, Masumoto Kouji
journal or publication title	Journal of Pediatric Surgery Case Reports
volume	3
number	12
page range	573-576
year	2015-12
権利	(C) 2015 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
URL	http://hdl.handle.net/2241/00137687

doi: 10.1016/j.epsc.2015.10.019

J Ped Surg Case Reports 3 (2015) 573–576

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

Gastric outlet obstruction caused by an ectopic pancreas in a neonate: A case report

Shunsuke Fujii [a], Hideaki Tanaka [a,*], Kentaro Ono [a], Yutaro Onizawa [b], Yayoi Miyazono [b], Naoya Sakamoto [a], Chikashi Gotoh [a], Yasuhisa Urita [a], Toko Shinkai [a], Hajime Takayasu [a], Ryo Sumazaki [b], Kouji Masumoto [a]

[a] Department of Pediatric Surgery, Faculty of Medicine, University of Tsukuba, 1-1-1 Tennoudai, Tsukuba, Ibaraki 305-8575, Japan
[b] Department of Pediatrics, Faculty of Medicine, University of Tsukuba, Tsukuba, Ibaraki, Japan

ARTICLE INFO

Article history:
Received 4 October 2015
Received in revised form
24 October 2015
Accepted 26 October 2015

Key words:
Ectopic pancreas
Neonate
Obstruction

ABSTRACT

We herein report a neonate who presented with non-bilious vomiting at one day of age caused by a prepyloric ectopic pancreas. Ultrasonography clearly detected the presence of a submucosal mass preoperatively, which was treated with local gastric resection. Only 9 neonates with a symptomatic pyloric or prepyloric ectopic pancreas have been previously reported in the literature. Therefore, we reviewed and discussed the clinical features of neonates with this type of ectopic pancreas.

Ectopic pancreas is defined as pancreatic tissue that lacks anatomic and vascular continuity with the main body of the pancreas [1]. It is commonly asymptomatic and incidentally found in the stomach, duodenum or upper part of the jejunum during operations and at autopsy [2]. Symptomatic ectopic pancreas in neonates is extremely rare. We herein report a one-day-old female neonate who presented with non-bilious vomiting caused by an obstruction secondary to an ectopic pancreas located at the prepyloric antrum.

1. Case report

A 3125 g female baby, one of dichorionic diamniotic twins, was born at 37 weeks of gestation by cesarean section, indication for which was a previous cesarean delivery. Her prenatal surveys had revealed normal amounts of amniotic fluid. Her Apgar scores were 8 and 9 at 1 and 5 min after birth, respectively. Her oral intake was started soon after birth as usual, however, she presented with non-bilious vomiting 4–6 times per day on the first day after birth. She had been conservatively observed in the newborn nursery until six days after birth. A physical examination at that time revealed a soft abdomen and normal bowel sounds without a palpable mass. The patient's body weight was 2768 g, which showed an 11.4% decrease compared to her birth body weight. Her blood tests were unremarkable except for mild hyperbilirubinemia. An abdominal X-ray demonstrated a gas in the large intestine predominantly and did not indicate a complete bowel obstruction. Abdominal ultrasonography (US) was then performed, which revealed a submucosal solid mass (measuring approximately 7 mm in diameter) at the anterior wall of the prepyloric gastric antrum (Fig. 1). The width of the pyloric wall and length of the pyloric channel were within normal limits, which excluded hypertrophic pyloric stenosis (HPS). An upper gastrointestinal (UGI) series demonstrated a narrow pylorus and a round-shaped defect of the contrast medium at the prepylorus in the prone position, potentially due to the above mentioned mass (Fig. 2). Thus, a jejunal tube was placed for enteral feeding. Contrast-enhanced abdominal computed tomography (CT) was performed to confirm the characteristics of the mass and it localized the slightly enhanced mass at the same site indicated on US. The preoperative diagnosis was a submucosal mass at the prepyloric gastric antrum, which included the differential diagnoses of ectopic pancreas and gastric duplication. At 16 days of age, the infant underwent exploratory laparotomy via an upper half circumumbilical incision. The mass was palpated at the anterior

* Corresponding author. Tel.: +81 29 853 3094; fax: +81 29 853 3091.
E-mail address: tanaka-h@md.tsukuba.ac.jp (H. Tanaka).

http://dx.doi.org/10.1016/j.epsc.2015.10.019

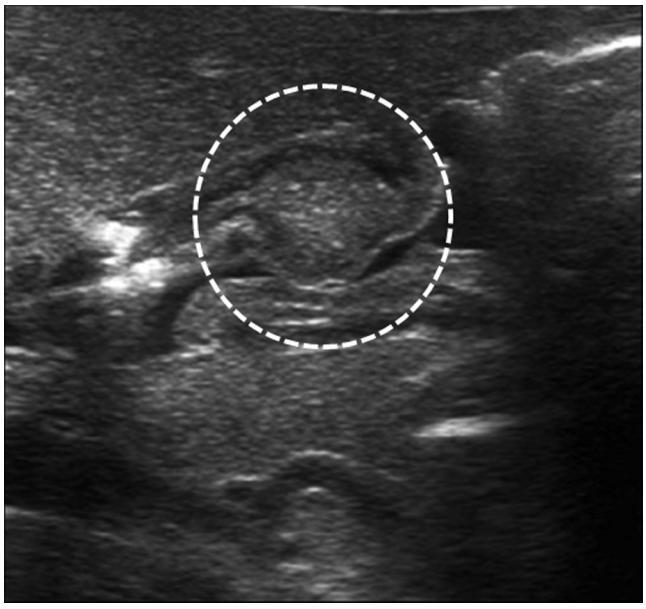

Fig. 1. Abdominal ultrasonography at the age of 7 days revealed submucosal mass at the anterior wall of prepyloric antrum (dotted circle).

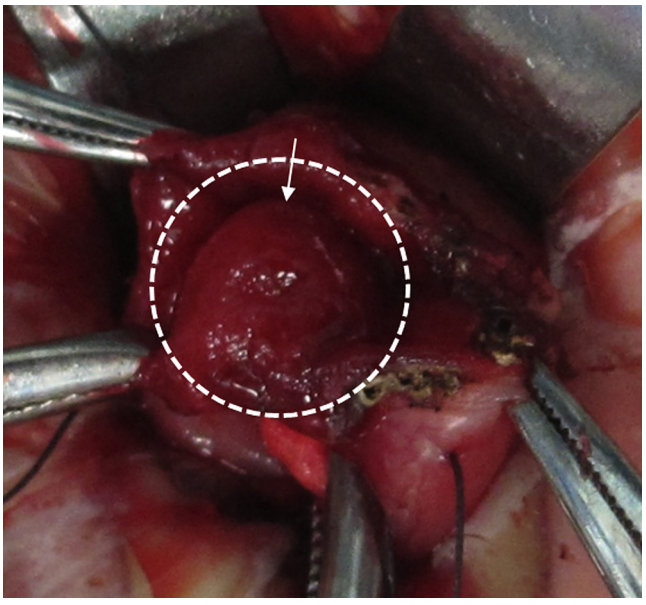

Fig. 3. Intraoperative finding: part of the anterior wall of prepyloric antrum was incised adjacent to the base of the submucosal mass (dotted circle), and the central umbilication was noted on the surface (arrow).

wall of the antrum and excised with a full thickness of the local gastric wall (Fig. 3). The defect of the antral wall was closed in a single layer with interrupted sutures. It had a central umbilication on the mucosal surface of the mass. The postoperative course was uneventful except for slow recovery of gastric emptying. She was discharged on the 24th postoperative day and has gained normal growth and development during the follow-up period of 18 months. The excised mass specimen consisted of a $10 \times 10 \times 8$ mm, solid, milk-white colored tissue. A histologic examination revealed aberrant submucosal exocrine pancreatic tissue that had acini, ducts, and islets cells, which was compatible with an ectopic pancreas (Fig. 4).

2. Discussion

Ectopic pancreas occurs in 1%–2% of autopsies and in 1:500 laparotomies with a male to female ratio of 3:1 [1]. Ninety percent of all cases are found in the stomach, duodenum, and jejunum, although it can be found throughout the gastrointestinal tract and in other intra-abdominal organs [2]. The majority of patients with ectopic pancreas are clinically asymptomatic. Its clinical manifestation may include some degree of obstruction resulting from the enlarged nodule or complications such as inflammation, ulceration, tumor, or intussusception [3].

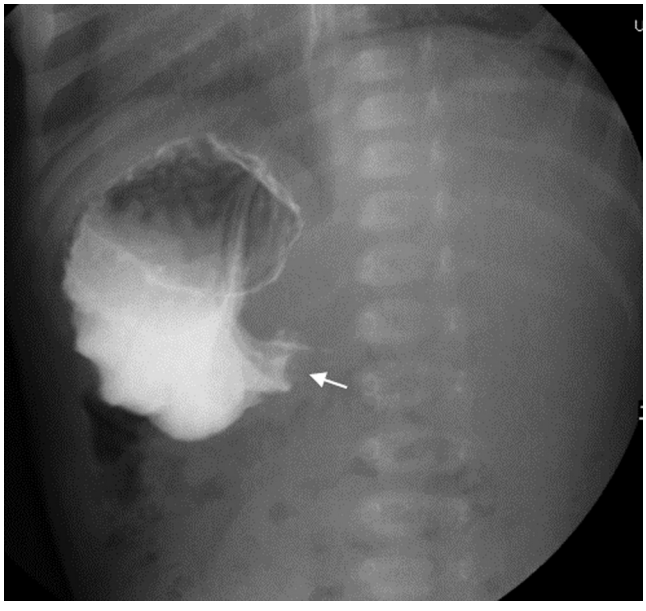

Fig. 2. Upper gastrointestinal series demonstrated narrowed pylorus and a round-shaped defect of the contrast medium in the prone position at the prepylorus (white arrow).

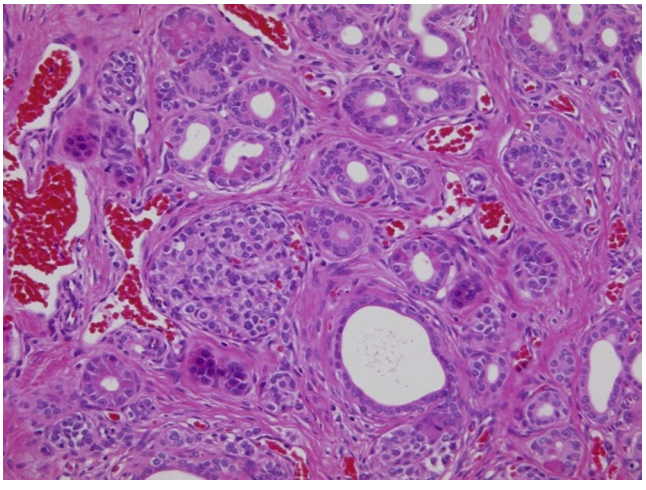

Fig. 4. Histologic section of the submucosal mass (Hematoxine eosine staining): pancreatic tissue with acini, ducts, and islets cells was noted in the layer of between submucosa and muscularis propria.

Table 1
Neonatal cases with symptomatic ectopic pancreas located at pylorus or prepyloric antrum.

Patient no	Author	Year	Sex	Onset	Symptom(s)	Preoperative diagnosis	Preoperative imagings for diagnosis	Operation	Age at operation	Time of final diagnosis	Outcome	Size
#1	Kernohan [4]	1956	M	10 days	Non-bilious vomiting	HPS	UGI series	Ramstedt's operation	20 days	At autopsy	Died of intestinal bleeding	NA
#2	Matsumoto [5]	1974	F	5 days	Non-bilious vomiting and melena	HPS	Physical exam	(1) Gastrojejunostomy (2) Anterectomy, gastroduodenostomy	(1) 7 days (2) 6 months	After 2nd operation	Alive	NA
#3	Ishihara [6]	1990	F	20 days	Non-bilious vomiting	HPS	UGI series	(1) Wedge resection (2) Antrectomy	(1) NA (2) 8 days after 1st operation	After 1st operation	Alive	3 mm
#4	Visentin [7]	1991	M	18 days	Non-bilious vomiting	Hypertrophic pyloric mass	US	(1) Biopsy, gastrojejunostomy (2) Detach the gastrojejunostomy, antral mass untreated	(1) 18 days (2) 2 yo	Biopsy	Alive	NA
#5	Ueno [8]	1993	F	10 days	Hematemesis and melena	Submucosal tumor	Endoscopy, UGI series	Tumor resection	6 months	After operation	Alive	NA
#6	Hayes-Jordan [9]	1998	M	2 days	Non-bilious vomiting	Submucosal tumor	UGI series, endoscopy, US	Subserosal excision	3 days	After operation	Alive	3-4 mm
#7	Ozcan [10]	2002	M	7 days	Non-bilious vomiting	Pyloric stenosis, or HPS	UGI series, US	Subserosal excision	1 mo	After operation	Alive	4-5 mm
#8	Fragoso [11]	2004	M	21 days	Non-bilious vomiting	Ectopic pancreas	Endoscopy, US	Partial gastrectomy	40 days	After operation	Alive	NA
#9	Surov [12]	2009	F	12 days	Hematemesis and melena	Arteriovenous malformation	US, CT	NA	NA	After operation	Alive	NA
#10	Our case	2015	F	1 day	Non-bilious vomiting	Ectopic pancreas, gastric duplication	US, UGI series, CT	Resection of tumor with local gastric wall	16 days	After operation	Alive	10 mm

HPS: hypertrophic pyloric stenosis, CT: computed tomography, UGI: upper gastrointestinal contrast study, US: ultrasonography, NA: not available.

To the best of our knowledge, only 9 neonatal cases with symptomatic ectopic pancreas located at the pylorus or prepyloric antrum have been previously reported in the literature [4–12]. Table 1 shows the demographics and clinical outcomes of these cases, including ours. The onset of the symptoms ranged from one day to 21 days of age. Most of the initial symptoms were vomiting. Gastrointestinal bleeding was noted in three cases (Pt#2, 5, 9). The clinical courses of the first four cases (Pt#1–4) were not straightforward; Pt#1 had Ramstedt's operation without treating the lesion and subsequently died of gastrointestinal bleeding. Pt#2 and #3 had two operations because the primary operations only revealed the pyloric mass, which necessitated antrectomy later in secondary operations. Pt#4 underwent gastrojejunostomy and a biopsy of the prepyloric lesion in the primary operation, revealing the ectopic pancreas. The physicians decided to observe the patient without removing the lesion and had the anastomosis detached, thereby regaining normal intestinal continuity in the secondary operation. A submucosal tumor at the prepyloric lesion was detected by endoscopy in three cases (Pt#5, 6, 8), and a central umbilication on the mucosa was also noted in Pt#5. An UGI series was performed in six cases (Pt#1, 3, 5, 6, 7, 10); a prepyloric mass was demonstrated in three cases (Pt#5: prepyloric mass with a central umbilication, #6 and #10: antral mass), and partial or complete pyloric obstruction was the only finding in the remaining cases. When the imaging study was limited to the UGI series, a correct diagnosis was hard to obtain preoperatively (Pt#1, 3, 7). Abdominal US was able to detect the prepyloric mass in four cases (Pt#4, 6, 9, 10), leading to additional imaging studies on the suspicion of pathologies other than HPS in three cases (Pt#6, 9, 10).

HPS is most often encountered as a cause of non-bilious vomiting in early infancy, and a physical examination and abdominal US, revealing a palpable enlarged pylorus and hypertrophied pyloric muscle with elongated pyloric channel, respectively, are common diagnostic methods for HPS [13]. If these examinations do not confirm HPS, then the differential diagnoses include various conditions such as pylorospasm, gastroesophageal reflux, gastric volvulus, antral web, preampullary duodenal stenosis, duplication cyst, and ectopic pancreas [13]. Abdominal US remains important for the differential diagnosis because it may visualize the ectopic pancreas as a pyloric or prepyloric mass, such as in our case. The UGI series may visualize the submucosal mass with central umbilication, but may only show a narrowed pylorus and may not be consistent with an ectopic pancreas. Endoscopy would be able to visualize the submucosal mass with a central umbilication as a typical feature of the ectopic pancreas, however, it requires experienced pediatric gastroenterologists or surgeons who are familiar with this procedure under general anesthesia for neonates. CT with contrast enhancement may detect the ectopic pancreas as a mass and may rule out other abnormalities, but its merit should be weighed against its risk of radiation exposure. If abdominal US clearly detects the submucosal mass around the gastric outlet, then that would be adequate for diagnosis. A correct diagnosis and precise localization of the ectopic pancreas would avoid unnecessary operations or overtreatment, thereby leading to its local excision with a minimal incisional approach, such as a circumumbilical incision as was performed in our case or a laparoscopic approach.

References

[1] Fam S, O'Brian D, Borger J. Ectopic pancreas with acute inflammation. J Pediatr Surg 1982;17:86–7.
[2] Doran R, ReMine W, Dockerty M. The fate of heterotopic pancreatic tissue. Arch Surg 1974;109:762–5.
[3] Feldman M, Weinberg T. Aberrant pancreas: a cause of duodenal syndrome. J Am Med Assoc 1952;148:893–8.
[4] Kernohan R, Morison J. Symptomatic pancreatic heterotopia of the pylorus associated with bilateral renal cortical necrosis in an infant. Arch Dis Child 1956;31:276–8.

[5] Matsumoto Y, Kawai Y, Kimura K. Aberrant pancreas causing pyloric obstruction. Surgery 1974;76:827–9.
[6] Ishihara M, Yamazaki T, Ariwa R, Imura S, Maezawa M. Aberrant pancreatic tissue causing pyloric obstruction. Pediatr Surg Int 1990;5:140–1.
[7] Visentin M, Guitard V, Juskiewenski S. Pyloric obstruction with complicated pancreatic heterotopia. Eur J Pediatr Surg 1991;1:247–8.
[8] Ueno S, Ishida H, Hayashi A, Kamagata S, Morikawa M. Heterotopic pancreas as a rare cause of gastrointestinal hemorrhage in the newborn: report of a case. Surg Today 1993;23:269–72.
[9] Hayes-Jordan A, Idowu O, Cohen R. Ectopic pancreas as the cause of gastric outlet obstruction in a newborn. Pediatr Radiol 1998;28: 868–70.
[10] Ozcan C, Celik A, Guclu C, Balik E. A rare cause of gastric outlet obstruction in the newborn: pyloric ectopic pancreas. J Pediatr Surg 2002;37:119–20.
[11] Fragoso A, Correia-Pinto J, Carvalho J, Dias J, Troncoso M, Estevao-Costa J. Ectopic pancreas and foveolar hyperplasia in a newborn: a unifying etiopathogenesis for gastric outlet obstruction. J Pediatr Gastroenterol Nutr 2004;39: 92–4.
[12] Surov A, Hainz M, Hainz M, Holzhausen H, Finke R, Spielmann R, et al. Ectopic pancreas with pseudocyst and pseudoaneurysm formation. Clin Radiol 2009; 64:734–7.
[13] Fujimoto T. Infantile hypertrophic pyloric stenosis. In: Puri P, Hollwarth M, editors. Pediatric surgery: diagnosis and management. Berlin Heidelberg: Springer-Verlag; 2009. p. 363–8.